

Case Reports

Systemic *Streptococcus equi* Infection in a Horse Handler—A Case of Human Strangles

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SERIOUS INFECTIONS with group C streptococci are uncommon in humans. Of the four species in this group, *Streptococcus equisimilis* has most frequently been reported to cause infections in humans.¹ These have included bacteremia, meningitis, endocarditis and pneumonia.² Another species within this group, *Streptococcus equi*, is found most often in horses and is the causative agent of strangles, an acute, contagious and deadly respiratory tract disease that attacks young horses particularly and has led to devastating epidemics in stables where many horses are housed.³ There is only one reported case of human infection with *S equi* in a woman with bacteremia who had undergone multiple grafting procedures after a radical vulvectomy for carcinoma.⁴ We report a case of bacteremia and cellulitis due to *S equi* in an ostler (horse handler or caretaker).

Report of a Case

A 56-year-old man was admitted to hospital with a four-day history of increasing swelling and pain over the right side of his face. He had experienced fevers, night sweats and rigors for two days before admission. The facial swelling had begun at the angle of the right mandible and had progressed to involve the right parotid and submaxillary region. On the evening before admission the patient noted a decrease in the facial swelling but redness and swelling had spread into the neck and anterior chest area. Two weeks before admission, a dry cough developed that had not resolved at the time of admission. There was no history of previous significant illness. The patient had worked for the past 20 years as a caretaker of horses, providing them with food and water and cleaning the stables. He lived in a mobile home within 3 m (10 ft) of a stable. None of the horses at that stable had recently been ill. He smoked two packs of cigarettes per day and drank 6 to 12 beers per day.

On physical examination, the patient was disheveled but appeared in no distress. The temperature was 36.8°C (98.2°F). The right side of the face was erythematous, warm

and considerably swollen from the inferior border of the right orbit encompassing the right parotid, submaxillary and mandibular regions and the right neck and suprasternal notch area where there was tender fluctuance (Figure 1). Erythema and mild induration were also noted over the anterior chest and abdomen extending to the inguinal ligaments bilaterally. There was no crepitation at any site. The patient was edentulous except for two lower incisors that were carious but not tender to percussion.

The leukocyte count was 21,200 per μ l. The blood urea nitrogen level was 18 mg per dl, the creatinine was 1.5 mg per dl and the urine showed 20 erythrocytes per high power field, 10 to 15 leukocytes per high power field and no casts, with 300 mg per dl of protein. The fluctuant area over the suprasternal notch was aspirated and Gram-positive cocci in chains and Gram-negative rods were seen. Culture of the aspirate from the wound and two blood cultures grew a β -hemolytic *Streptococcus* that was sensitive to bacitracin. The organism was identified as group C by Lancefield antiserum and as *S equi* by its nonfermentation of trehalose, lactose and sorbitol. The minimal bactericidal concentration of penicillin was 0.025 μ g per ml and 4 μ g per ml of gentamicin. *Bacteroides oralis* and *Bacteroides melaninogenicus* grew from the wound as well.

The patient was treated with penicillin and required surgical drainage of 50 ml of purulent fluid from the fascial planes of the right side of the face. Culture of this fluid also grew *S equi*. The patient's condition gradually improved over the following two weeks and the cellulitis had resolved three weeks after admission. The creatinine level rose to 2.5 mg per dl in the first week of hospital stay but subsequently decreased to 1.2 mg per dl on discharge from the hospital. Results of the urinalysis became normal.

Comment

In young horses, strangles can be fatal and occur in epidemic form where many horses are housed. Infected horses initially are quiet and lethargic and there is fever, rhinitis and pharyngitis. Subsequently, there is swelling of cervical nodes, which can drain purulent material. Bacteremia occurs frequently, and pneumonia and meningitis can occur. The disease is transmitted by inhaled or ingested nasal discharge. The microorganisms can remain viable in contaminated feed or water troughs for weeks.³ A carrier state exists among horses, and, in one case, a horse continued to shed *S equi* for ten months after an initial mild infection, despite being asymptomatic during that period.⁵ Protection by means of immunization is incomplete⁶ and yearly boosters are recommended.⁷

Our patient had worked with horses for 20 years and lived in close proximity to a stable. The portal of entry of the organism may have been through his mouth via one of his remaining incisors, which were carious. That an infection with this organism should develop in him is remarkable and suggests an overwhelming inoculum of bacteria. His upper

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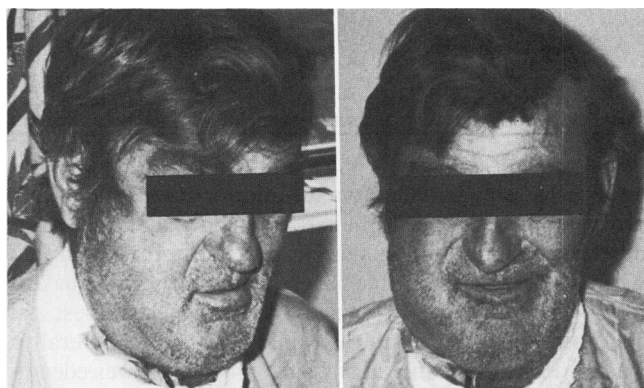


Figure 1.—The patient three days after surgical drainage. **Left**, view from right side; **right**, frontal view.

respiratory tract symptom of dry cough, subsequent development of a facial cellulitis with bacteremia with *S equi* and spread of pus along fascial planes to the suprasternal notch as well as induration of the skin of his chest and abdomen to the level of the inguinal ligament are strikingly similar to the syndrome that occurs in horses and, as such, may represent the first described case of "human strangles."

Of interest also is the initial evidence (and subsequent resolution) of renal disease reflected by microscopic hematuria, pyuria and proteinuria in the presence of a rising creatinine level. The development of poststreptococcal glomerulonephritis has been associated with infections with another group C *Streptococcus*, *Streptococcus zooepidemicus*.⁸ In this case the initial respiratory tract infection two weeks previously may have served as an immunologic stimulus for the development of nephritis. *S equi* does not produce streptolysin O; thus antistreptolysin O would be expected to be negative.

Group C streptococci are most frequently β -hemolytic on blood agar but γ - and α -hemolytic strains have been reported.⁹ When β -hemolytic streptococci are isolated, many laboratories test for susceptibility to bacitracin to separate group A β -hemolytic streptococci, which are routinely sensitive to bacitracin, from other β -hemolytic streptococci. Of group C streptococci, however, 6% to 71% have been noted to be susceptible to bacitracin,¹ as was the organism in this case. Therefore, in most circumstances, testing of β -hemolytic streptococci for bacitracin susceptibility should not substitute for grouping of the organism by Lancefield antiserum.

Penicillin has been the primary antimicrobial for treating group C streptococcal infections in general. It appears that ampicillin, cephalothin sodium, chloramphenicol, clindamycin and vancomycin hydrochloride may be acceptable alternatives.^{2,10} The issue of tolerance of group C streptococci to penicillin has been raised recently. In one study by Portnoy and co-workers,¹¹ 16 of 17 clinical isolates of group C streptococci showed a 32-fold or greater difference between minimal inhibitory and minimal bactericidal concentrations. Synergy between penicillin and gentamicin was found in all of the 17 strains. At some variance with this is the more recent study by Royston and associates¹⁰ that showed penicillin tolerance in only 2 of 26 cases. Nonetheless, in those situations wherein penicillin tolerance can be shown, the addition of an aminoglycoside may be advisable. In this case, the minimal bactericidal concentration was 0.025 μ g per ml; thus, the

patient was treated with penicillin without gentamicin in addition to surgical drainage to which the infection eventually responded.

S equi is one of four species of group C streptococci. Of the four, *S equisimilis* and *S zooepidemicus* are known to occasionally produce infection in humans. *Streptococcus dysgalactiae* affects cows and sheep and is a rare human pathogen. This is the second report of *S equi* infection in a human but the first of a syndrome resembling strangles.

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Myopathy From Surreptitious Ipecac Ingestion

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EMETINE HYDROCHLORIDE, the major alkaloid constituent of ipecac, has clinically recognized neuromuscular toxicity.¹⁻⁶ Acute and subacute to chronic toxic myopathies have been described, but reports of human biopsy specimens are uncommon.^{2,4-6} In recent reports the reversibility of this uncommon toxic myopathy primarily in association with abuse of ipecac syrup has been emphasized.^{3,4,6}

We studied the case of a 27-year-old woman with a history of progressive proximal muscle weakness and elevated muscle enzyme levels. Results of the muscle biopsy were abnormal and the specimen showed irregular inclusions in both type 1 and type 2 muscle fibers. It was later discovered that she had been surreptitiously ingesting large amounts of ipecac syrup for several months to lose weight. We review previous reports of ipecac myopathy, emphasizing the clinical features and findings on muscle biopsy. Clinicians should be alerted to this toxic myopathy, especially in young women with a possible eating disorder.

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